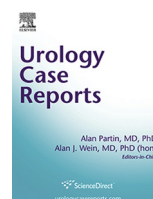


Contents lists available at ScienceDirect

Urology Case Reports

journal homepage: www.elsevier.com/locate/eucr

Trauma and Reconstruction

Massive Renal Intratubular Hemorrhage With Herniations into Renal Veins: Report of a Case

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ARTICLE INFO

Article history:

Received 15 November 2014

Accepted 25 November 2014

Available online 20 December 2014

Keywords:

Haematuria

Intratubular hemorrhage

Tamm–Horsfall protein

Histopathology

ABSTRACT

A 30-year-old woman suffered intractable gross hematuria of renal origin, which required nephrectomy after failed conservative management. Pathological examination revealed massive intratubular hemorrhage and frequent deposition of an amorphous and homogenous material positive for Periodic Acid-Schiff (PAS) stain in the corticomedullary junction. This substance is frequently herniated into the lumen of thin-walled veins of arcuate size. There was no apparent cause for the bleeding. To the best of our knowledge this is the fourth reported case with these peculiar histopathological findings associated with hematuria. We discuss some hypotheses as to the etiopathogenesis of this rare and interesting condition.

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Case report

A 30-year-old Asian woman was presented with total gross hematuria and left flank pain.

She denied trauma or urinary lithiasis. She suffered UTI 2 years ago, treated with oral antibiotics. She is single and non-smoker. Physical examination was unremarkable. A complete blood study was normal. Cytological study was not conclusive. A urine culture was negative. Axial computerized tomography (CT) revealed enlarged left kidney with decreased uptake of contrast, no masses could be seen (Fig. 1). Urethrocystoscopy revealed multiple blood clots in the bladder, hematuric urine efflux from the left ureter. The bladder mucosa was normal.

A left ureteroscopy revealed blood clots in the renal pelvis and unremarkable mucosa. Selective urinary cytology was negative. After conservative management the hematuria did not resolve multiple blood transfusions were needed. Radionuclide scan (DMSA) demonstrated poorly functioning left kidney. We proceeded for left nephrectomy. Intra-operatively, left kidney was congested with adhesions to the surroundings and 75% of the surface of the kidney had a mottled tan and dark red appearance. The renal artery was normal. The renal vein had no thrombosis. No lesions were noted within the collecting system.

On the cut surface extensive dark areas compromised the cortex, including the columns of Bertin. The medulla and some cortical area

were spared. The pelvis and ureter were unremarkable. No thrombi or other lesions were noted in the renal artery and vein. No vascular malformations or tumors were found.

Microscopically, the most striking finding was a massive intratubular hemorrhage with tubular dilatation in the cortex (Figs. 2 and 3).

Frequently, the blood was hemolyzed and was in the form of homogenous protein casts. There was slight hemorrhage in the medulla with no tubular dilatation. In some areas atrophic tubules with interstitial fibrosis and a slight focal mononuclear cell infiltrate were seen. There was a sharp limit between hemorrhagic and normal cortex. In the corticomedullary junction there was frequent deposition of an amorphous and homogenous material that was intensely positive for Periodic Acid-Schiff stain. A unique finding was the presence of this material close to thin-walled veins of arcuate size frequently herniating into the lumen. Some of these intravenous polyps were covered by fibrin.

Discussion

Solez and Heptinstall demonstrated that the amorphous material in the interstitium or herniating into the veins at the corticomedullary junction was Tamm–Horsfall protein.¹ This glycoprotein, present in normal urine, is the primary constituent of urinary casts and is localized selectively along surface membranes of the thick ascending loop of Henle.²

One of the most frequent causes of extravasation of Tamm–Horsfall protein in the interstitium is increased intratubular

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Figure 1. Axial CT scan shows enlarged left kidney with dilated pelvis and decreased contrast uptake.

pressure as in obstructive uropathy.³ This protein also can herniate into the renal veins.

In the third case reported by Iliff and Galdabini the intra hemorrhage could be due to the intrarenal vascular aneurysm but in their cases 1 and 2, and in our own, no apparent cause was detected.¹ Billis, Palma, Prando and Gouvea theorized that the intratubular hemorrhage might be caused by the venous blood gaining access to the tubular lumens by way of the tubulovenous communication.

If this is correct it is hard to understand why intratubular hemorrhage does not occur more frequently, taking into account the frequency of these communications. A backflow of blood under high pressure from the pelvis into the tubules seems to be improbable because there was little blood in the medulla and the collecting ducts in this area were not dilated.

They also mentioned another possibility that the hemorrhage was from the glomeruli but this is also improbable because there was no blood in Bowman's space and all of the glomeruli appeared normal. The distribution of the intratubular hemorrhage involving segments of the cortex suggests that the bleeding originating most probably from the vessels of corticomedullary junction.⁴

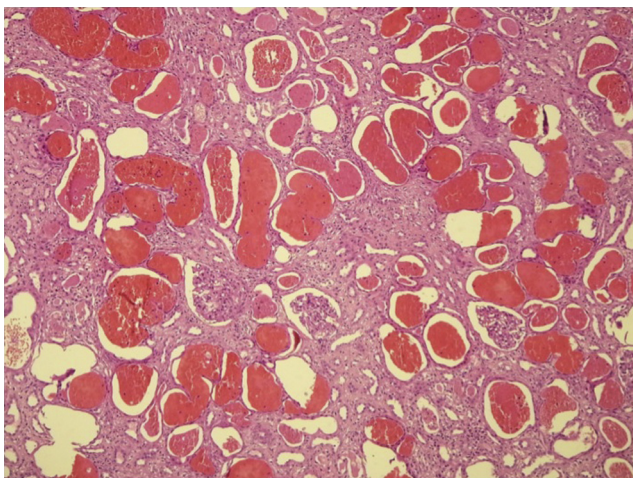


Figure 2. Massive intratubular hemorrhage with dilation (H&E stain).

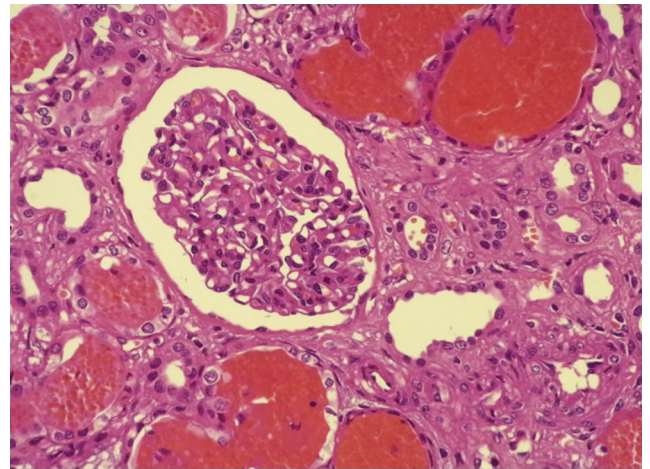


Figure 3. Higher power view of the slide shown in Fig. 2.

The only cases in which Solez and Heptinstall noted Tamm–Horsfall protein in Bowman's space were those with massive tubular hemorrhage.¹ According to these investigators this finding could be interpreted as indicating retrograde flow in the tubules and, by analogy, the red cells in the tubules originated lower down in the nephron, namely at the sites of the ruptures at the cortico-medullary junction.

A possibility worth mentioning is rupture of the fornical venous plexus.⁴

Billis, Palma, Prando and Gouvea speculate on a hypothetical sequence of events in cases 1 and 2 reported by Iliff and Galdabini⁵ that the primary event leading to hematuria could be a ruptured vessel at the fornical venous plexus.⁴

Vesicoureteral reflux could be another possibility but is less likely because the urine culture was negative and the configuration of the ureteral orifices was normal during the cystoscopic examination. Acute hydronephrosis due to the blood clots at the pyelocaliceal system may increase the intratubular pressure, with extravasation of Tamm–Horsfall protein in the interstitium and into the veins forming tubulovenous communications.⁴

The 3 patients reported on by Iliff and Galdabini were cured after nephrectomy⁵ as well as our patient.

We hope that new cases will be able to clarify the etiopathogenesis of this condition.

Conflict of interest

No conflicts of interest for any of the authors.

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